



## ABSTRACT

Pudendal neuralgia is a form of chronic pelvic pain, although the validity of this diagnosis has not been firmly established. It is characterized by unilateral pain in the pudendal nerve dermatome, mostly while sitting. In this case report, a 76-year-old female patient presented to her general practitioner with a 10-year history of unilateral buttock pain, which had been diagnosed as pudendal neuralgia. She tried various treatments and visited numerous specialists, but there was little improvement overall. Some medications seemed to have a positive effect, but pain ultimately persisted until psychological factors were appropriately addressed. Management of pudendal neuralgia follows general chronic neuralgia principles, from medication over nerve block to definitive surgery. However, this case serves as a prime example of how the psychosocial approach is equally important. Given that psychological factors can make pain persist, it is imperative to properly address them. As pudendal neuralgia is uncommonly encountered in primary care, specific attention must be paid to psychological factors, which become significantly more important in long-standing chronic pelvic pain.

**KEYWORDS:** Pudendal neuralgia, primary care, holistic approach

# Pudendal Neuralgia: The Need for a Holistic Approach—Lessons From a Case Report

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Chronic primary pain (CPP) is defined as pain lasting more than three months, with significant emotional and psychological disability.<sup>1</sup> CPP is frequently encountered in primary care and its management is often a challenge.<sup>2</sup> In addition to the physiological basis of the pain itself, the psychological aspects often complicate matters.<sup>3</sup> Pudendal neuralgia (PN) is a form of pelvic CPP, classically characterized by unilateral pain in the distribution of the pudendal nerve (e.g., buttocks, perineum, and genitals).<sup>4</sup> The validity of this diagnosis, however, remains disputed.<sup>5</sup> Current treatment options begin with conservative management, followed by neuropathic pain medication and local infiltrations. Lastly, surgical nerve release is an option in refractory cases.<sup>4</sup>

Hereafter, we describe a case of long-standing PN in primary care, in which the current pharmacological approaches failed when the psychological needs of the patient were not adequately addressed.

## CASE

A 76-year-old woman presented to her general practitioner (GP) in late 2018 with a 10-year history of a continuous sharp, burning pain, situated in the left buttock, perineum, and vagina. The pain started after a maneuver performed by an osteopath she visited for chronic back problems. Following a referral to a pain clinic after her initial presentation in 2008, she was diagnosed with PN.

The pain was more pronounced when sitting down or lying on the affected site and

significantly less intense when standing or walking around. She had no pain when sitting on a toilet seat, there was no dyspareunia, and sleep was rarely interrupted. The condition had had a severe impact on her daily functioning, straining her marriage and social life. Her medical history was significant for previous trigeminal neuralgia (2008), for which she was effectively treated with valproate, before the onset of PN. She had a previous episiotomy for the birth of her daughter in 1975, and her family history was positive for fibromyalgia (sister).

After the PN diagnosis, she was first treated with physiotherapy and classical analgesics. Additionally, the patient was treated with duloxetine, gabapentin, and amitriptyline, with minimal to mild reduction in pain, but subsequently discontinued because of intolerable side effects. Topical lidocaine patches were also used, but they were ultimately found impractical. At presentation to her GP, her regimen was pregabalin 300mg in the evening, tramadol 100mg as needed (up to 3 times a day) and 10 clonazepam drops (2.5mg/mL) at night. She felt these medications helped her sleep better, but they provided minimal pain reduction.

From 2010 to 2013, the patient received numerous steroid infiltrations in the left ischial tuberosity, with limited and mostly temporary results. In 2014, pulsed radiofrequency was used, without any pain relief. There was even a short trial with a neurostimulator in early 2018, without any positive effect. She had a psychiatric consultation before the

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implantation of the neurostimulator, where no contraindications for stimulator placement were withheld.

A pelvic magnetic resonance imaging (MRI), performed in 2013, did not reveal abnormalities of the pudendal nerve, and an ultrasound of the hip region in 2014 revealed significant chronic enthesopathy of the hamstrings. Because of this, she was given a course of high-dose ibuprofen at the time, primarily to reduce inflammation. This did not have any effect on the pain. Gynecological examinations and ultrasounds were normal. Psychological evaluation, performed at a pain clinic in 2010, showed a cluster B personality with histrionic features and little insight.

The initial clinical interview was marked by her occasionally getting up from her seat and standing upright. Speech was hyperverbal and circumstantial, and she frequently appeared anxious. When seated, she sat only on the right buttock; her gait showed no abnormalities; the sensitivity, strength, and mobility in her back and limbs were all normal; reflexes were normal; there was significant tenderness when palpating the left ischial tuberosity. In the following consultations, the patient sometimes brought up family conflicts, especially with her husband and daughter, as she felt they did not take her or her condition seriously. Moreover, she appeared sad when discussing this issue.

She was started on a trial of carbamazepine (200mg twice daily), which was eventually discontinued because of agitation and dizziness. Palmitoylethanolamide (PEA) was then introduced at 1200mg (i.e., 400mg three times daily). This initially had a positive influence on the vaginal symptoms: there was a reduction in the hypersensitivity of the clitoral region, which she described as a tingling, burning sensation.

A psychological referral was suggested by her GP, but she repeatedly refused psychological treatment, insisting that there was solely a physiological basis for her pain. At her request, a trial of cannabis was started. Surgery was offered, but she opted to wait.

Ultimately, she had undergone a coccygectomy, with no effect on the pain. During the next year, she had appointments with her GP every two weeks. She received education on how pain is modulated by emotion and the complexity of chronic pain in general, and her troubled relationships with her husband and daughter were addressed as well. She

realized she held a negative attitude toward her pain and the perception of her condition by her immediate family. Consultations now focused more on acceptance and education about psychological factors involving pain (including catastrophizing), rather than medical or surgical treatment of her PN. No new treatment was started, but she was instead advised to continue physiotherapy and her initial medication (pregabalin, tramadol, and clonazepam). After approximately one year, her attitude toward her condition had changed, and her overall quality of life improved. She was less anxious in clinical interviews and sat down more and for longer periods of time. She reported that her pain was still there, although less markedly. More importantly, she acknowledged that her own perception and behavior toward her pain, as well as the family conflicts, might have contributed to the pain severity, while also recognizing her own responsibility regarding her situation.

## DISCUSSION

In our view, this case highlights some important aspects of chronic pain as we see it in primary care. First, there is the diagnosis itself. The existence of PN as a proper diagnosis rather than a syndrome has been contested,<sup>5</sup> and the available literature mostly consists of case reports. The exact prevalence remains unknown. It can be caused by acute nerve injury following trauma, as presumed in this case, but other risk factors have been described: pelvic surgery or childbirth (particularly with episiotomies), mechanical nerve entrapment by muscle spasm or cicatrization, postherpetic neuralgia, and even cycling.<sup>4,6</sup>

Pain is limited to the distribution of the pudendal nerve (anus, perineum, penis/scrotum, or vagina/clitoris). It typically occurs unilaterally, mostly when sitting on a hard, flat surface, which leads to compression of the nerve. Pain is frequently completely absent while standing or lying on the unaffected side or even, as in our case, when sitting on a toilet seat with no direct pressure on the nerve.<sup>4,7</sup> This specific presentation has led to the development of the Nantes criteria, aiding physicians in making the diagnosis of PN: a) pain in the pudendal nerve dermatome, b) pain mostly while sitting, c) no pain at night, d) no sensory impairment, and e) pain relief by pudendal nerve block.<sup>7</sup> All these criteria are considered essential.

Diagnosis on clinical grounds alone, such as in a GP's office, is technically not possible, if adhering strictly to these criteria. A positive response to a nerve block remains essential. However, this procedure can also be therapeutic,<sup>6</sup> and as such it seems reasonable to diagnose on clinical grounds first after ruling out other pathology. Conservative therapy can already be started before referring the patient for a nerve block, which simultaneously confirms the diagnosis.

As already stated, it is important to rule out other pathologies, such as chronic hamstring tendinopathy or malignancy. A full genital/gynecological examination is reasonable. An ultrasound and MRI of the pelvic and hip area can reveal possible inflammatory processes or anatomic anomalies of the pudendal nerve. Similarly, lumbar spine pathology should be ruled out by CT or MRI.

Management of PN follows the principle of chronic pain treatment in general. Conservative treatment is first-choice and remains important throughout. This therapy can be easily initiated in primary care. There are various pharmacological options, which should be chosen in function of patient tolerance and clinical response, which might take a few trials.<sup>8</sup> Amitriptyline, duloxetine, and the antiepileptics pregabalin and gabapentin are the best studied for neuropathic pain. In experimental settings, PEA has been effective in PN.<sup>9</sup> There was some degree of pain control in our patient as well. In any case, response to treatment should be evaluated frequently and management adjusted accordingly.

Ischial tuberosity infiltrations for pudendal nerve block can be considered after failure or inadequate pain resolution with conservative therapy. Surgical referral for pudendal nerve release should be offered as a last resort. Patients who have had a positive response to a pudendal nerve block are ideal candidates.<sup>10</sup>

The existing literature on trigeminal neuralgia, a condition similar in many aspects to PN, might provide some further insight into appropriate management. It typically occurs on one side of the face in the distribution of cranial nerve V and is most often described as a stabbing, piercing pain.<sup>11</sup> The course of the condition is interspersed with periods of remission, but it generally tends to become less responsive to therapy over time, with eventually surgery being required. Long-standing pain

and longer attack duration are unfavorable prognostic factors.<sup>11,12</sup>

However, as illustrated in this case, chronic pain is complex and can involve many more elements than just aberrant conduction in nerve tissue. Without a doubt, chronic pain conditions can have a devastating impact on patients' personal and social lives. Our patient attended little to no social events and barely had family or friends over because she could not sit for extended periods of time.

Besides the fact that there is a genetic component to chronic pain,<sup>13</sup> psychological factors also play an important role, particularly in its persistence. Relationships with family members, especially those with a chronic pain syndrome, can influence pain perception and behavior.<sup>2</sup> Furthermore, a negative attitude towards pain ("catastrophizing") can predict its severity in a sample of patients with postherpetic neuralgia.<sup>14</sup> A similar effect was seen in patients with spinal injury.<sup>15</sup> This of course does not necessarily imply causation, but tailoring treatment to address these psychological mechanisms might prove useful, as it appears to do in fibromyalgia.<sup>16</sup> There is even some evidence that the central nervous system processing of pain in these patients is altered, contributing to pain chronicity.<sup>17</sup> During initial clinical interviews, our patient demonstrated an emotional, negative, and sometimes even dramatizing attitude toward her pain.

In this particular case, our patient was diagnosed with cluster B personality disorder at a psychological assessment in the pain clinic. Cluster B personality disorders, particularly of the borderline type, have been associated with an increased risk for chronic pain.<sup>18</sup> The heightened emotional sensitivity and dysregulation underlying these disorders is thought to contribute to pain processing, making psychological interventions significantly more important in these patients.<sup>18,19</sup>

## CONCLUSION

Research suggests that using a holistic approach to treat difficult chronic pain syndromes, such as PN, is more beneficial to the patient than focusing only on physiologic aspects of the disease.

We can try to eliminate pain by conservative and invasive measures, medication, and physiotherapy. However, taking this case as

a learning opportunity, it is useful to also address feelings of hopelessness or depression in patients. This includes providing education in how they view pain negatively, how it contributes to pain processing, and how to work towards changing their attitudes. Psychological support might help, but must be proposed carefully, with consideration for the patient's own views and preferences. This multimodal approach, incorporating both medication, invasive techniques, and psychological support, has already been proposed for long-standing trigeminal neuralgia.<sup>20</sup> This multifaceted approach allowed our patient to eventually have an acceptable degree of pain control and a better quality of life.

The longer chronic pain is present the less contributory value a specific diagnosis has, as it does not help guide treatment at that point forward. A holistic and effective approach for such longstanding chronic pain can be achieved by addressing all possible social and psychological factors as well as the underlying physiological mechanisms.

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